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New Finding of Multiple Inflamed Follicular Cysts in Two Patients With CANDLE Syndrome: A Case Report

Tyler Holley
University of Nebraska Medical Center

Melissa Moutray
Private Practice

Indranell Bhattacharyya
University of Florida

Chad S. Sloan
University of Nebraska Medical Center

Valmont P. Desa
University of Nebraska Medical Center

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Conclusion: In one of the largest studies of prostate cancer in RA, we estimated prostate cancer rates to be similar in a national Veteran population with RA to those in the general population. The small difference may be accounted for by cases not captured within ORD. This study was limited by the indirect standardization of prostate cancer rates to SEER. There is need for future research with direct standardization to US Veterans. ■

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New Finding of Multiple Inflamed Follicular Cysts in Two Patients With CANDLE Syndrome: A Case Report

Tyler J. Holley¹, Melissa Moutray², Indranell Bhattacharyya³, **Chad S Sloan¹**, Valmont P Desa¹

¹University of Nebraska Medical Center, Department of Surgery, Division of Oral and Maxillofacial Surgery

²Oral and Maxillofacial Surgeon, Private Practice, Garden City, KS

³University of Florida, Department of Pathology, Division of Oral and Maxillofacial Pathology, Gainesville, FL

Mentor: Valmont P Desa

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Background: CANDLE Syndrome (Chronic Atypical Neutrophilic Dermatositis with Lipodystrophy and Elevated Temperature) is an autoinflammatory disease. Characteristics include: recurrent fevers, organ inflammation, skin lesions, anemia, lipodystrophy, basal ganglion calcifications, and delayed physical development. Orofacial manifestations include facial lipodystrophy, swollen eyelids, thick lips, macroglossia, generalized microdontia, and jaw osteopenia. We describe novel cases of two patients, diagnosed with CANDLE syndrome, who presented with radiolucent mandibular lesions associated with displaced permanent molars and mandibular bony expansion.

Methods: Patient 1: A 9-year-old male with CANDLE Syndrome was referred to our clinic for evaluation of a left mandibular radiolucency. After appropriate work-up, the lesion was surgically removed, found to be cystic in characteristic, measured 2x2x0.5cm and filled with a dark-brown turbid fluid. Two-years later, the patient returned with complaint of movement of his teeth. Repeat imaging showed a recurrent 3x2x1cm lesion.

Patient 2: A 10-year-old male with CANDLE Syndrome was referred to our clinic for

radiographic findings of a right mandibular radiolucency. Computed tomography imaging showed a 4x3x1cm lesion and an additional 1x1x1cm lesion of the left mandibular body (Figure 1).

Results: Histopathologic analysis of all four lesions revealed cysts of follicular odontogenic origin with intense inflammatory cell infiltrate.

Conclusions: To our knowledge, there has not been a previously described case of patients

with CANDLE Syndrome presenting with inflammatory odontogenic follicular cysts. The inflammatory cell infiltrate coincides with the inflammatory dysregulation of disease and organ inflammation seen globally. The new finding of radiolucent mandibular cystic lesions provides further insight into the extent and characterization of the Syndrome. ■

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Figure 1. Orthopantomogram demonstrating right and left lesions of the mandible with associated displaced mandibular right second molar and mandibular left first molar, respectively. Additional CT imaging identified right posterior mandibular lesion dimensions of 4x3x1cm and left mandibular body lesion of 1x1x1cm.